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Case Report

"CAVERNS" of vessels in a male breast: a rare site for hemangioma

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Abstract

Incidence of male breast lesions has increased by 1.6% in the last two decades. Nevertheless, hemangioma of breast is seldom encountered in clinical practice (0.8%) and its incidence in male breast is unknown due to extreme rarity. A 72 year old male, with 40 years history of slowly progressing, painless, mass in the left breast, mammography showing an oval lobulated dense mass with fine calcifications and FNAC yielding only hemorrhage underwent wide local excision. Histopathological diagnosis was in favour of cavernous hemangioma. Rarity of male breast lesions pause a diagnostic and prognostic challenge due to little evidence available in the literature. Imaging and cytology being nonspecific, in large lesions differentiation from the malignant counterpart often requires complete excision and histo-morphological examination, aided with ancillary technique like IHC.

Keywords: Hemangioma breast, Male breast.

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1. Introduction

Incidence of male breast lesions has increased by 1.6% in the last two decades.[1] Conditions that affect female breast are encountered in male breast as well.[2] Nevertheless, hemangioma of female breast is seldom encountered in clinical practice (0.8%) and its incidence in male breast is unknown due to extreme rarity.[3,4] Almost all palpable vascular tumours of male breast have been hemangiomas. Cavernous hemangiomas are one of the most common forms of mammary hemangiomas and when picked up, must be distinguished from more ominous lesions.[5,6] Herein, we report a case of 72 year old male with palpable breast mass, histo-pathologically diagnosed as cavernous hemangioma.

2. Case Report

A 72 year old male presented with a 40 years history of slowly progressing, painless, large palpable mass in the left breast, involving all quadrants equally. He was a known case of hypertension, hypothyroidism and asthma and was on regular medications. As an initial diagnostic approach, the patient was referred for mammography. Diagnostic mammography showed an 11.2 x 7.4 x 6 cm oval lobulated dense mass with fine calcifications. Subsequent FNAC could yield only hemorrhage. Hence with a provisional diagnosis of a possible vascular tumour, patient was subjected to wide local excision considering the large size of the tumour and the specimen was later sent for histopathological examination.

We received a skin covered grey brown spongy mass measuring 11.5 × 7 × 8cm. The overlying skin was unremarkable. On sectioning, grittiness was felt and micro cystic spaces were noted on cut surface (**Figure 1**A). Representative bits were submitted. Microscopic examination revealed skin with an underlying ill-defined lesion composed of multiple dilated vascular channels of varying sizes. These were lined by flattened to plump endothelial cells. Areas of papillary fronds, thrombosis and calcification were also noted (**Figure 1** B,C). The intervening stroma was scant and showed chronic mononuclear inflammatory infiltrate. All these features histo-

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morphologically favoured cavernous hemangioma. However, immuohistochemistry was performed for Ki 67 proliferative index to rule out the possibility of any overt angiosarcoma in the background of a large ill-defined lesion with the presence of papillary fronds. Ki 67 immunostaining was performed using monoclonal antibody MIB-1 and appropriate staining protocol was used with lymph node tissue as internal control. Conventional eye- balling method derived a KI 67 proliferative index of 0-1% (**Figure 1**D). This confirmed the diagnosis of a benign vascular tumour.

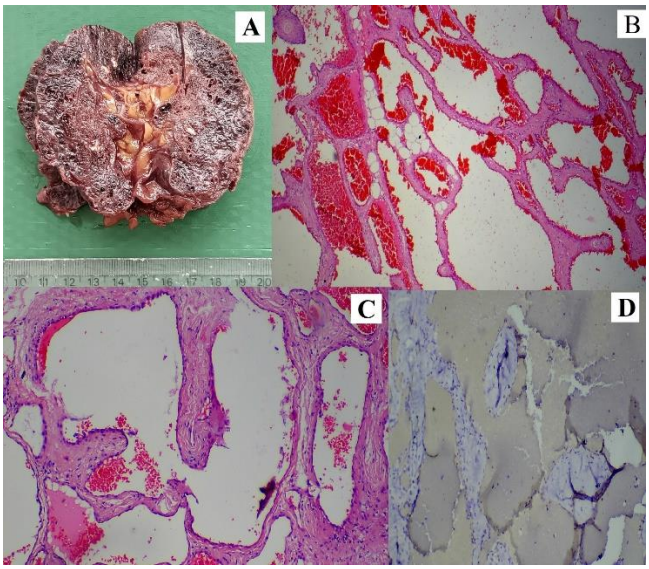

Figure 1: (**A-D**): **A):** Gross photograph of a grey brown spongy mass with multiple tiny cystic spaces. **B**): Dilated vascular channels lined by endothelial cells (H&E; 10x). **C):** Papillary fronds into the vascular channel (H&E; 40x). **D):** IHC staining for Ki67 shows 0-1% of the lining endothelial cells exhibiting nuclear positivity

3. Discussion

Hemangiomas are benign vascular tumours most commonly presented in infancy and childhood, often involving head or neck and liver internally.[6] As described by Rosen et al, in breast, hemangiomas can be subcutaneous non parenchymal, parenchymal or perilobular. Parenchymal hemangiomas are further divided into cavernous, non-cavernous, capillary, complex or venous hemangiomas.[5] Cavernous hemangiomas are characterized by large elastic variably sized blood filled spaces lined by single layer of flat endothelial cells.[7]

In females, age distribution ranges from 19 to 82 years with mean age being 60 years.[5] Reviewing the online scientific database of available case reports on cavernous hemangiomas in male breast, the presenting age is between 52 to 84 years, including our case. The duration of presenting illness ranged from few days to several months to several years, as in the present case. The latter would probably be due to the painless nature of the lesion in 80% of reported cases. The average size of the tumour in females range from 0.3 to 2.5 cm, whereas males presented with lesions of larger size ranging from 0.7 to 14cm in diameter. Hence when only 40% of females present with a palpable mass, almost all of the male breast hemangiomas had a palpable mass or nodule.[4]

Recent studies could elicit the possible mechanism of development of these vascular tumours and are attributed to abnormal cytokines or pathways involving factors like hypoxia- inducible- factor- 1α (HIF- 1α), platelet derived growth factor receptor- beta (PDGFR-β) and vascular endothelial growth factor (VEGF). Downstream pathways including phosphatidylinositol 3 kinase (PI3K) / protein kinase B (Akt) are also involved which results in the abnormal migration and proliferation of vascular endothelial cells and thereby developing hemangiomas.[8]

Mammographically in both male and female hemangiomas can be round or oval masses with well delineated margins and may contain fine or coarse calcification.[9] Calcification is a non-specific finding and does not point toward malignancy. Fine needle aspiration usually are paucicellular with few benign mesenchymal cells in a hemorrhagic background.[4] Complete excision is required for an accurate diagnosis.

Grossly cavernous hemangiomas are well circumscribed, firm and dark brown spongy mass. Microscopically dilated vessels lined by endothelial cells with inconspicuous flat nuclei, congested with red blood cells and supported by fibrous stroma are seen. Calcification occur in the stroma. Borders might drift into surrounding breast parenchyma and organizing thrombi might show papillary endothelial hyperplasia. Also the mostly independent channels might show anastomosis at areas.[5] All these findings tend the histopathologists to consider angiosarcoma as a close differential.

The need for extreme caution in the interpretation of grossly evident vascular tumours is always emphasized, because portions of angiosarcomas may appear deceptively bland histologically. Similarly cavernous hemangioma can duplicate the histologic appearance of peripheral parts of some well differentiated angiosarcomas. Extra-medullary hematopoiesis, if present, is an important diagnostic clue for the benign nature.[5] Literature has also shown evidences of transformation of benign hemangiomas to malignant angiosarcomas of low grade, though rare. Factors like prolonged duration of the lesion, exposure to radiations or certain exogenous hormones are seen to influence most of these hemangioma- angiosarcoma transition. Nevertheless few case reports could not elicit any such factors. Unlike carcinomas, the precursor malignancies and the tumorogenesis are still being explored in soft tissue lesions.[10-13] The advent of proliferation marker Ki 67 and its immunostaining in such instances plays a role in differentiating the malignant counterpart. In the present case the Ki 67 proliferative index was 0-1%. Ki 67 proliferative index rarely exceeds 5% in benign hemangiomas, where as in angiosarcomas they are invariably >20%.[5] This has a proven

sensitivity and specificity of 90% and 95% respectively in breast vascular tumors alone.[14]

4. Conclusion

Rarity of individual male breast lesions pause a diagnostic and prognostic challenge due to little evidence available in the literature.[15] Diagnostic imaging and cytology being nonspecific, in large lesions differentiation from the malignant counterpart often requires complete excision and histo-morphological examination, aided with ancillary technique like IHC.

5. Source of Funding

None.

6. Conflict of Interest

None.

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